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CASE REPORT

Surgical management of chronic traumatic pseudomeningocele of the craniocervical junction: case report

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Abstract
Purpose Chronic traumatic pseudomeningocele (PM) is a rare complication of gunshot injuries of the craniocervical junction in pediatric patients. Impairment of the CSF dynamics may cause severe symptoms and should be treated.
Methods We report the case of a 6-year-old girl who was accidentally shot in the neck during tribal clashes. On being admitted, she was neurologically intact with cerebrospinal fluid (CSF) leakage through the wounds. She underwent primary closure of the wounds in a rural medical facility. After two episodes of meningitis, CSF leakage resolved spontaneously. Nine months later, the patient was presented with a disfiguring mass growing in the posterior neck, severe headaches, and constitutional symptoms such as loss of appetite and a failure to thrive.
Results Neurosurgical intervention was performed with the patient in the prone position. Occipital pericranium graft was used to repair the defect, and the cavity of the PM was obliterated with muscle layers. The patient's symptoms improved at 1 year follow-up without PM recurrence.
Conclusion This is a rare presentation of gunshot injuries in an environment with limited neurosurgical resources. Restoring the normal pattern of CSF circulation should be the aim of any neurosurgical intervention.

Keywords Pseudomeningocele · Craniocervical junction · CSF fistula · Intracranial hypotension

Introduction

Gunshot injuries are common in developing countries. There is currently a trend in tribal societies to use guns during clashes, which not infrequently results in significant morbidity or death in affected individuals [7, 8, 21]. Gunshot injuries can involve the central nervous system and result in devastating neurologic damage, vascular injury, and meningitis [1, 8, 20, 21]. Traumatic pseudomeningocele (PM) is a rare complication of gunshot injury to the spine that can be managed by conservative measures and cerebrospinal fluid (CSF) diversion in the earliest stages [6, 17]. Conservative management is therefore usually ineffective in the chronic stages of PM after a cavity is established, with resulting intracranial hypotension and CSF hypovolemia [10]. Here, we report a case of chronic traumatic PM of the craniocervical junction (CCJ) in severely symptomatic child with severe symptoms due to a neck mass effect and impairment CSF dynamics. Surgical repair of the traumatic pseudomeningocele resulted in a symptom-free child.

Case report

A 6-year-old girl was involved in tribal clashes and was injured laterally by a gunshot in the posterior aspect of the neck at the CCJ level. The patient remained neurologically intact and clear fluid was leaking from the wound upon reporting to the hospital. The patient underwent primary

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closure of the wounds in a rural medical facility. Three days later, the wound became infected and CSF leakage from the wound was evident. One month later, after two episodes of meningitis, CSF leakage improved spontaneously. Nine months later, the patient was readmitted for a disfiguring pulsating mass on the neck, severe lingering headaches, loss of appetite, and failure to thrive during the last 6 months. A head computed tomography (CT) scan showed a PM of the CCJ widely communicated with the cisterna magna (Fig. 1). On examination, the patient was alert with reactive pupils. Furthermore, cranial nerve examination revealed no abnormalities, including no evidence of paresis or hypoesthesia in the limbs. After a thorough evaluation of the case, we decided to repair the defect due to the lack of improvement 9 months after the injury. With the patient in prone position and the head fixed with the three-point Mayfield-Kees fixation device, a midline skin incision was performed from 4 cm above the occipital protuberance to the C2 vertebrae (Fig. 1). We performed a careful dissection of cervical soft tissues and muscles. A T-shaped incision was made at the fascial plane and muscles. Then, a subperiosteal dissection of the cervical spine was carried out in order to expose the area from occipital squama to the spinous process of the C2 vertebrae. Thus, we gained access to the whole occipitocervical area which was occupied by a large cavity full of CSF. The cyst was penetrated and the fluid drained, allowing us to obtain good visualization of the defect between the inferior border of the foramen magnum and the superior border of the posterior arch of C1. The extensive cavity had no free dural edges, and the posterior aspect of the medulla oblongata was entirely exposed (Fig. 2).

To repair the dural defect, an occipital 6×3 cm pericranium graft was obtained and sutured to the border of the defect with silk stitches at a 3 mm interval. To obliterate the dead space, the three muscular layers were reapproximated with sutures over the pericranium graft covering the defect. The fascial plane then was closed with interrupted sutures. Finally, the skin was closed with vertical mattress stitches. At the immediate postoperative period, the patient was neurologically intact without CSF fistula. Eight months later, the patient remained asymptomatic without PM (Fig. 2). Due to financial issues, the patient has not been able to obtain a control CT scan of the head. However, due to the absence of symptoms and no PM recurrence, control imaging studies are not mandatory for this patient, mainly in developing countries with healthcare services costly for patients.

Discussion

We present a case of traumatic PM as a result of gunshot wound in the neck that caused severe symptoms during

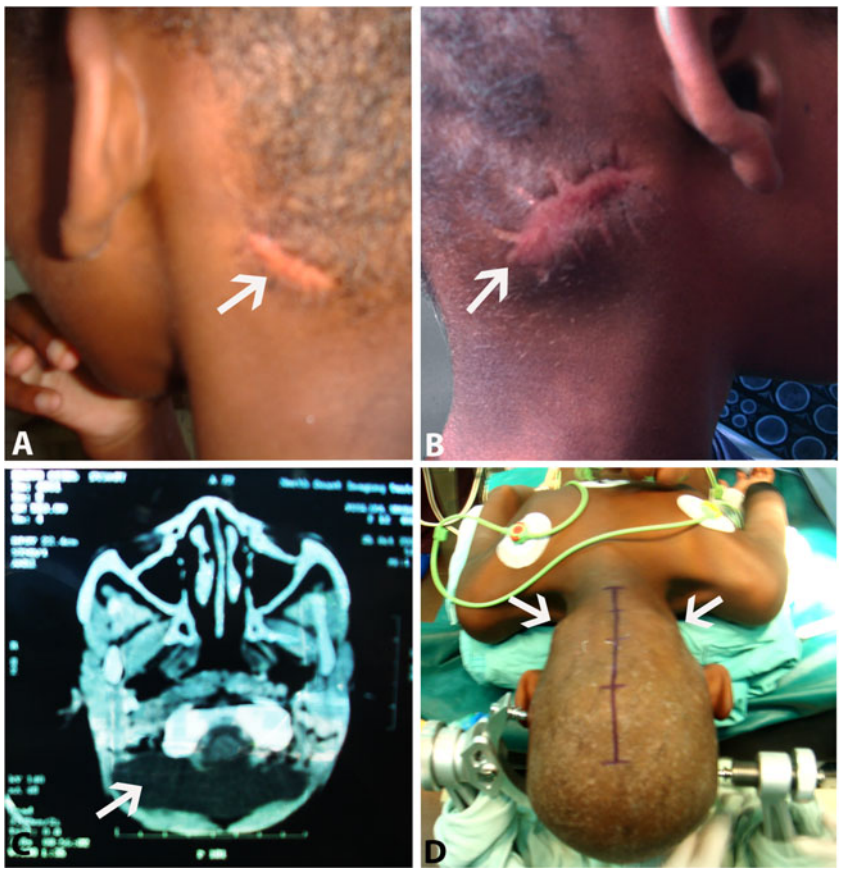

Fig. 1 Photograph of the child with traumatic PM. **a** The scars of the penetrating injury in the posterior aspect of the neck are shown with the typical smaller entrance wound of the bullet and **b** larger exit wound. **c** CT scan of the head showed the PM (*arrow*). **d** The child was operated in prone position with midline incision 4 cm over the occipital eminence to the C2 vertebrae. We can also see the swelling of the scars due to PM (*arrows*)

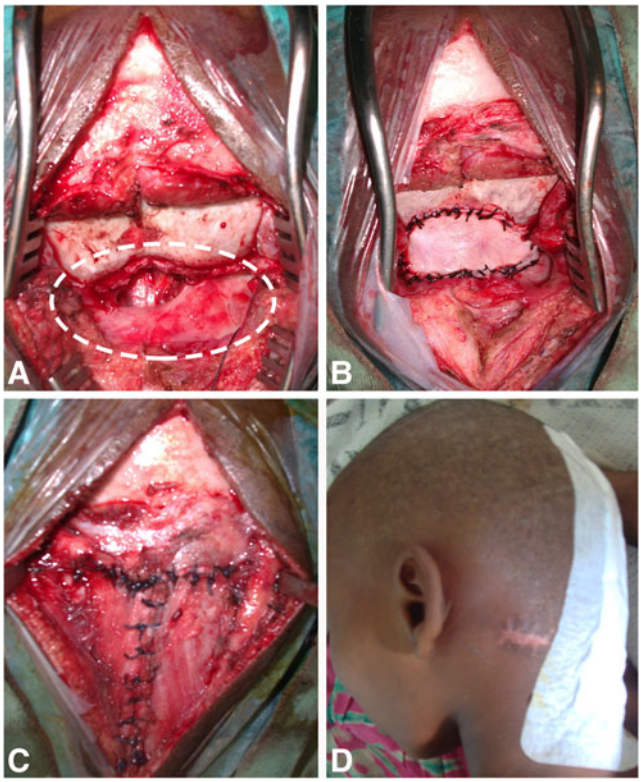

Fig. 2 Intraoperative photos. **a** The *circle* shows the extent of the PM. The medulla oblongata was entirely exposed. **b** Occipital pericranium flap was used for reconstruction; the graft was sutured around the defect. **c** The muscular layers were opened in a T-shaped fashion, and they were closed upon the graft to obliterate the cavity. **d** 5 days later, the child was asymptomatic without CSF fistula

6 months. Impairment of CSF dynamics was the main cause of her clinical manifestations, and restoring the normal CSF circulation was the aim of the surgical approach despite of a situation with limited neurosurgical resources. Regarding the mechanism of the injury, the bullet went laterally through the posterior cervical muscles with a small left lateral entry zone and a bigger right lateral exit wound due to the conic effect of the bullets [1, 15]. The bullet passed below the posterior rim of the foramen magnum and the upper surface of the C1 posterior arch piercing the dura but leaving intact the cervical spinal cord, which led to the formation of a big intramuscular cavity of the CSF [4]. Usually, this type of injury may cause severe damage to the spinal cord. Indirect injuries may also occur due to fractures and dislocations of the spine, and the outcome will depend on the spinal cord damage [18]. The initial management of this type of injury will require immediate debridement of the tissues. The evident bullet fragments should be carefully removed. Infection, hematoma, and persistent CSF leakage should also be managed aggressively to avoid secondary damage of the spinal cord [1, 17, 18].

Penetrating trauma of the cervical spine is usually associated with spine instability and vascular injuries. To address this, the appropriate workup should be carried out with angiography and endovascular treatment if necessary. CT scan of the injured spine segment could be important to evaluate the need for surgical intervention [21]. On the other hand, when there is no neurologic deficit, infection, or CSF leakage, conservative management is recommended [3].

The most common traumatic PM is due to brachial and lumbosacral plexus injury [6]. Atlanto-occipital dislocation may cause secondary PM due to dural lacerations in the zone of the trauma that are associated with neurologic compromise in some cases [5, 11, 12, 16]. In this setting, it has been reported that hydrocephalus and syrinx are associated with the PM [11]. These pathologic associations highlight the concept of an alteration of the intracranial dynamics due to an ectopic cavity filled with CSF acting as a large reservoir that produces a decrease in intracranial pressure and impairment of the CSF reabsorption in the arachnoid granulations, which act as pressure-dependent valves driven by the cardiorespiratory rhythm [2, 10]. The mechanism by which the CSF is contained in the cavity of the pseudomeningocele remains unclear. It has been hypothesized that a ball-valve mechanism in the site of the dural laceration may play a role in the persistence of the cavity [12]. Some authors argue that PM results from arachnoid-intact herniation through the dural defect; this phenomenon is referred to as "true meningoceles" [6]. CSF collections may tend to be spontaneously reabsorbed in the soft tissue, but when a cavity is established, the CSF becomes more difficult to reabsorb and less likely to be resolved with conservative treatment. In our case, we did not find any dural laceration; however, we did find a large cavity filled with CSF that had developed, with neovascularization in the walls widely connected with the cisterna magna.

There are many strategies to treat CSF disorders associated with a PM. Direct dural repair is not always possible because the location of the dural tears may not be readily accessible for a given surgical approach. In the acute setting, traumatic PM could be treated with conservative measures, such as bed rest and acetazolamide, or with more aggressive management such as a blood patch, subarachnoid external drainage, or a lumboperitoneal shunt. These interventions are usually quite effective in the early stages of a PM when dural tears are apposed, and the aim of the intervention is to decrease the pressure of CSF pulsations favoring spontaneous closure [5, 11, 12]. Some authors have reported severe long-standing headaches which are recalcitrant to conservative treatment in patients who have been undergone lumbar puncture. When an occult CSF fistula is diagnosed, these authors advocate surgical repair of this type of dural injury to eliminate the headaches [13, 19]. Otherwise, an epidural blood patch is one of the strategies for use in acute or subacute stages of a PM that has been reported as an effective approach in many types of CSF collections; it remains one of the first invasive measures of choice when a PM is present [14].

When a long-standing PM becomes an encapsulated CSF cavity producing severe symptoms of intracranial hypotension, surgical repair is indicated with the aim of sealing the defect and eliminating the dead space to avoid recurrence [9]. Conservative measures may be ineffective due to integration of the pathological cavity with CSF spaces in the intracranial compartment. In this case, it is insufficient to only relieve pressure from the defect since a chronic PM does not usually have closely apposed free dural edges that can close spontaneously [9]. The operative technique to repair the PM includes primary closure of the dural defect with or without graft depending on the size of the defect using 4.0 nonabsorbable sutures at a 2 to 5-mm interval [6]. Sealant and fibrin glue can be used when leakage persists despite adequate suture closure. In our case, this was neither possible nor necessary because of successful closure with sutures and the lack of non-autologous materials in hospitals with limited resources. The muscular planes should be closed in two or three layers to obliterate the dead space between the dural repair and soft tissues [6]. Drainage should be avoided because it could promote the persistence of communication between the intra- and extradural space and results in high risk of infection.

In summary, traumatic PM is a rare complication of gunshot wounds involving the CCJ. In the pediatric population, impairment of the CSF dynamics may cause severe symptoms and should be treated with the aim of restoring the normal pattern of CSF circulation to reduce the symptoms. In chronic stages, surgical treatment with obliteration of the cavity and closure of the defect is mandatory due to the high probability of failure through conservative or less aggressive measures.

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Conflict of interest None

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